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Human nasal rhinosporidiosis - A case report

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ABSTRACT

Rhinosporidiosis is primarily an infection of the nose caused by *Rhinosporidium seeberi*. Other sites that can be involved are conjunctiva, urethra, palate, tongue, epiglottis, larynx, trachea, bronchi, skin, vulva, and vagina. It is endemic in certain parts of India and Sri Lanka. The pathogen is difficult to grow in culture and Hematoxylin and Eosin staining helps in confirmation of the etiological agent. Surgery remains the mainstay of treatment. Here is a report aimed at documenting a 26-year-old female patient with nasal rhinosporidiosis.

Key words: *Nasal mass, Polyploid lesion, Rhinosporidiosis, Rhinosporidium seeberi*

Rhinosporidiosis is primarily an infection of the nose caused by *Rhinosporidium seeberi*, lower fungi of aquatic habitat [1]. The main mode of transmission of rhinosporidiosis is through the spores which enters the human body from dust, infected clothing, fingers, or swimming in stagnant water. The taxonomic classification of *Rhinosporidium* was debated for few decades and recently classified into a new clade, mesomycetozoa which includes pathogen of fish and amphibians [2]. The disease is uniquely distributed in certain places such as India, Pakistan, and Sri Lanka. In India, the disease is more commonly reported from Tamil Nadu, Kerala, Puducherry, Andhra Pradesh, West Bengal, and Chhattisgarh [3].

R. seeberi causes chronic granulomatous disease on the mucous membrane and the estimated prevalence was reported to be 1.4% [4]. The most common site of the occurrence of rhinosporidiosis is nose and nasopharynx in 70% cases and ocular lesions particularly of the conjunctiva and lacrimal sac in 15% cases [5]. Occasionally, a disseminated infection involving limbs, trunks, rectum, and external genitalia are also reported in 15% of the cases [5]. The present case is an attempt to document the incidence of this enigmatic pseudofungal disease from this region. This data will enable the public health department to design strategies that help to reduce the prevalence of this disease.

CASE REPORT

A 26-year-old female patient from Cuddalore district of Tamil Nadu attended the otorhinolaryngology Outpatient Department (OPD) of Government General Hospital, Karaikal with the complaints of nasal blockage, right nasal mass, and disturbances of smell from right nostril and frequent episodes of a dry cough for the past 2 months. She reported no history of constitutional symptoms or any other chronic illness such as diabetes, epilepsy, ischemic heart disease, thyroid dysfunction, hepatitis, or pulmonary tuberculosis or tooth extraction/dentures or any other surgeries.

The patient was thin built, quiet, alert, and well-oriented with mild tachypnoeic. The pulse rate was 100/min and the blood pressure was 112/60 mmHg. Rest of the vitals were normal. The appearance of the nasal mass was strawberry such as friable, easily mobile, non-tender, and erythematous and bleeds on contact. It was about 3 cm in diameter and arising from the lateral aspect of the right inferior turbinate. The contralateral nasal cavity, nasopharynx, and palate were normal. There was no enlargement of regional lymph nodes and no other remarkable signs were noted on examination.

The blood sample was sent to the laboratory for hematological examination and the blood parameters showed hemoglobin - 9.5 g/dL; white blood cells - 8000 cells/cumm of blood with neutrophils - 58%, lymphocytes - 38%, and eosinophils - 4%. Serological test for hepatitis B surface antigen and antibodies for human immunodeficiency virus were negative.

On examination, clinician had a high index of suspicion for nasal rhinosporidiosis based on deviated nasal septum with the right nasal mass. Thus, the right nasal mass was cleared completely by surgical excision under local anesthesia. The nasal mass was removed and sent to the microbiology and pathology laboratory. A 10% potassium hydroxide (KOH) wet mount of surgically removed tissue revealed the presence of several spherical sporangia containing multiple sporangiospores in different stages of the development under 40x magnifications, morphologically suggestive of rhinosporidiosis (Fig. 1). Histopathology using Hematoxylin and Eosin staining showed a polypoidal lesion lined by pseudostratified ciliated columnar epithelium. Subepithelium shows mucous gland and inflammatory cells. A large number of spores are seen in the double-walled sporangia and also in submucosa confirmed the diagnosis of rhinosporidiosis (Fig. 2).

The patient recovered remarkably well after surgery. The patient was discharged and asked to report to ear, nose, and throat (OPD) every 6 months for follow-up. Unfortunately, she did not return for the follow-up examination.

DISCUSSION

Rhinosporidiosis was first described by Seeber from Argentina [1]. It is a chronic granulomatous disease commonly affecting the mucous membrane of the nasopharynx, anterior nares, inferior turbinate, septum, or nasal floor. Rarely, subcutaneous lesions of lower limbs, abdomen, and back have been reported [6]. However, there are reports of the disease that spread to anatomically distant sites through hematogenous or lymphatic routes [2].

The maximum cases are reported from endemic countries such as Sri Lanka and India especially from coastal areas [3]. The disease has been extensively seen in South Indian places such as Madurai, Thanjavur, Kanyakumari of Tamil Nadu, Alleppey, Kottayam, and Trivandrum district of Kerala [7], with few reported cases from non-endemic areas of East Delhi, Uttar Pradesh, and Gujarat [2,8,9]. The factors such as warm climates due to high temperature and increased humidity create a good environment for spore formation and contribute to hyperendemicity in Tamil Nadu.

In the present case, route of transmission was not known. However, the patient was a resident of rural area from Tamil Nadu where the occupation is mainly cattle rearing and agriculture. However, the highest incidence of cases is reported among red sand workers [10]. Although stagnant water is also a main source of transmission as the pathogen has aquatic habitat, in spite of several hundreds of persons bathe in our country, only a few develop progressive disease. It was suggested that an existence of some predisposing factors in the host can lead to the chronic granulomatous condition. Other risk factors include contaminated soil or waterfowl or even working in contaminated agricultural fields as reported earlier [2].

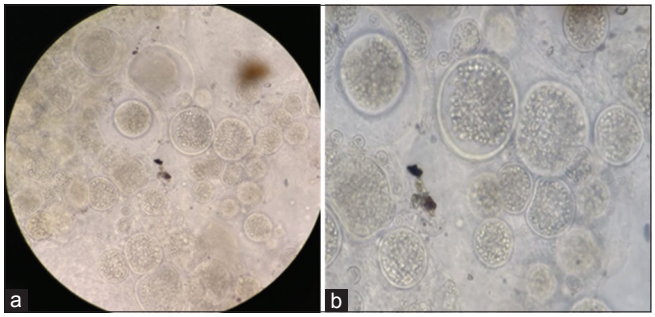

Figure 1: (a and b) 10% potassium hydroxide mount preparation (×400) showing numerous sporangia filled with endospores

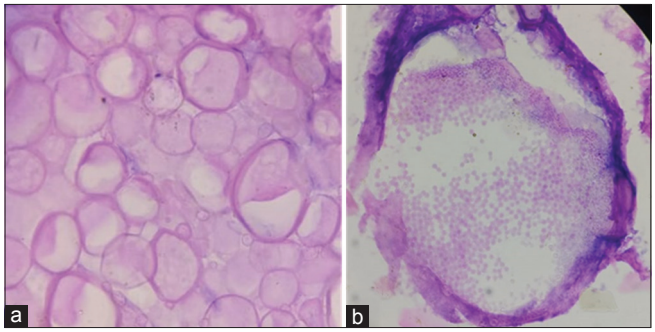

Figure 2: (a) Hematoxylin and Eosin (H and E) (×1000) shows numerous thick-walled sporangia; (b) H and E (×1000) shows a mature sporangium with numerous endospores in closer view

The disease is more prevalent in the second and third decade of life with a male to female ratio is 3:1 [3,10]. The present case correlates with the study where they stated that the highest incidence of rhinosporidiosis in India is more from blood Group "O" individual (70%), though occurrence in other blood groups are fairly equally distributed [5,8,10]. In contrast to this report, Jain stated that blood group is not a major factor to draw any conclusion [11].

Diagnosis of rhinosporidiosis mainly depends on microscopic findings of sporangia with sporangiospores, due to the failure of the microorganism to propagate by *in vitro* culture and the absence of a standard serological test for assessing the immune response of the host [2]. The characteristic morphological feature of spherical sporangium with numerous sporangiospores in a diverse stage of the development was noted under ×40 of 10% KOH wet mount (Fig. 1). Histopathology is customary for definitive diagnosis and could able to demonstrate double-walled sporangia with numerous spores (Fig. 2).

The present case had typical features of localized granulomatous strawberry like mass in the right nasal cavity made the clinician to have a high index of suspicion for rhinosporidiosis. The mass was excised under anesthesia. Conservative management with medical therapies such as dapsone or antifungals such as griseofulvin and amphotericin B showed limited results [12]. Although spontaneous regression is reported [12], the disease can be treated meticulously with complete and wide surgical excision followed by electrocautery of a basic polyp. Thus, the riddled organism with controversial taxonomy produces slow-growing masses in the nasal cavity that may confuse with soft tissue tumor.

CONCLUSION

Nasal rhinosporidiosis is endemic in India that resembles to that of neoplasm in its clinical features. Therefore, the clinician should consider nasal rhinosporidiosis in the differential diagnosis of a nasal mass.

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